

Case Report

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Parathyroid Adenoma Presenting as Spontaneous Cervical-Mediastinal and Retropleural Hematoma: A Case Report and Review of the Literature

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Abstract

Subcapsular hemorrhage is a well-known complication of parathyroid adenoma. However, extracapsular parathyroid hemorrhage can occur as a rare phenomenon. Here, we report the case of a patient with primary hyperparathyroidism manifesting as a cervical-mediastinal and retropleural hematoma and present an overview of previously reported cases.

Keywords: Parathyroid adenoma; Spontaneous hemorrhage; Hyperparathyroidism

Introduction

Sometimes, benign growths called adenomas appear on one or more of a person's parathyroid glands. Adenomas cause the parathyroid gland to make more parathyroid hormone than the body needs, a condition called primary hyperparathyroidism (PHPT). PHPT is a common condition in the United States, with a prevalence of approximately 1 case per 1,000 adults [1]. Too much parathyroid hormone increases the amount of calcium in the blood stream. However, hypercalcemia may not cause any symptoms. Parathyroid adenomas are usually discovered when a higher-than-normal calcium level shows up in a routine blood test [2].

Some people with mild or no symptoms of PHPT may decide to try hormone replacement therapy or medication options. However, surgery is the usual treatment for parathyroid adenoma, even for people who don't have obvious symptoms [2]. The patient reported had asymptomatic PHPT, which was not discovered until a hemorrhagic emergency. Here, we report a case of parathyroid adenoma manifesting as a cervical-mediastinal and retropleural hematoma and present an overview of previously reported cases. This type of presentation can be challenging even for an astute clinician.

A few cases of extracapsular hemorrhage caused by parathyroid adenoma have been reported. Most of them just gave a brief description of the problem. So we reviewed the relevant literature based on previous articles and summarized clinical details of available cases. Our study may help understand the clinical spectrum of the unusual presentation of common diseases. In case of spontaneous hemorrhage from an unknown origin, differential diagnosis of extracapsular parathyroid hemorrhage may be required.

Case Report

A 52-year-old woman was wounded in a fall. A chest radiogram revealed a rib fracture, and the patient was managed with conservative treatment. Her medical history was unremarkable, except for 2 rib fractures 16 years previously and a coccyx fracture at age 4. After a 10-day hospitalization, the patient presented with an unprovoked painful swelling and ecchymosis of the anterior neck. After 2 days, she presented with a massively enlarged neck, which was tender to touch (Figure 1). Her body temperature was 36.5°C, her blood pressure was 105/50 mmHg, and her pulse was 96 beats per minute. Laboratory tests revealed a hemoglobin level of 89 g/l, a serum calcium level of 2.92 mmol/l (reference range, 2.0-2.8 mmol/l), a serum phosphate level of 0.73 mmol/l (reference range, 0.8-1.4 mmol/l), and a parathyroid

hormone (PTH) level of 216.40 pg/ml (reference range, 15-65 pg/ml). Ultrasonography revealed an oval-shaped, hypoechoic lesion posterior to the left thyroid lobe, measuring 3.2×2.4×2 cm. And plenty of blood flow signals could be received from the lesion by color Doppler. Neck and chest computed tomography (CT) confirmed the presence of a massive cervical-mediastinal and retropleural hematoma (Figure 2). Ultrasound-guided aspiration yielded 15 ml of incoagulable blood from the retropleural space.

The patient was thought to have suffered a spontaneous extracapsular hemorrhage of a parathyroid adenoma. Supportive therapy was effective. The serum calcium levels remained high but stable during hospitalization, and the neck swelling gradually diminished (Figure 3). Repeated CT scans 10 and 24 days after presentation revealed progressive resolution of the hematoma. After 1 month of conservative treatment, we performed traditional bilateral open neck exploration. Nevertheless, the surgical procedure was difficult because of tissue adherence and was further complicated by the hematoma. An organized hematoma and 3 cm sized, dark brown colored cystic mass was found in posterior to the left thyroid lobe. The mass was dissected



Figure 1: **A)** Neck ecchymosis secondary to the extracapsular hemorrhage of parathyroid adenoma. **B)** The conservative treatment was effective, and the neck swelling and ecchymosis gradually diminished.

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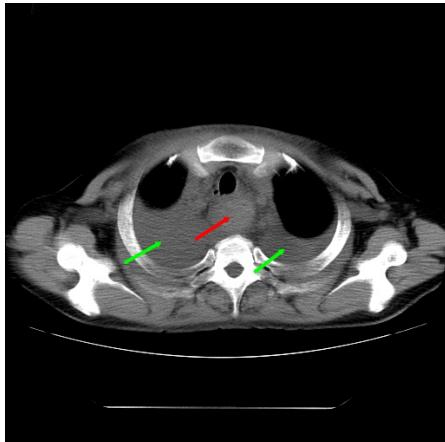


Figure 2: Computed tomography revealed a low-density mass in the cervical-mediastina (red arrow) and retropleural (green arrows) areas.

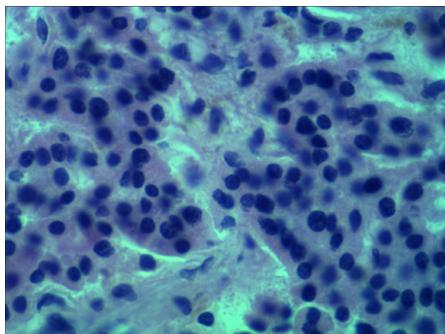


Figure 3: Hemosiderin deposition and fibrosis were consistent with a pathological diagnosis of parathyroid adenoma.

out with preservation of the recurrent laryngeal nerve. Final pathology revealed an enlarged parathyroid gland consistent with adenoma, with no evidence of malignancy (Figure 3). The postoperative course was unremarkable, and the patient made a full recovery with normalization of calcium, phosphate, and PTH levels within 3 days after surgery.

Discussion

The first case of spontaneous hemorrhaging of a parathyroid gland was reported by Capps in 1934 [3]. To date, 30 cases of extracapsular hemorrhage caused by parathyroid adenoma have since been reported [3-29]. Among these cases, 21 cases (70%) were in male patients, and 9 cases (30%) were in female patients. The mean age of the patients was 59.6 years (range 32-87 years). A review of these cases shows a wide range of age at diagnosis and slight male predominance.

The clinical presentations in the 30 cases varied and depended on the size and location of the hematoma and the PTH level. The usual presentation included hoarseness, dysphagia, dyspnea, cervical swelling, and cervical-thoracic ecchymosis. The serum calcium level was measured in 25 patients. Hypercalcemia was detected in 23 patients, and 2 patients had levels within or slightly lower than the normal range. The PTH level was measured in 15 patients, and, in almost all cases, the PTH level was elevated. Because the thyroid gland has a thick capsule, hematomas associated with thyroid glands are usually intracapsular. Intraparathyroid hemorrhage escapes through the capsule and produces a hematoma in the adjacent tissues. Hematomas

may extend into the mediastinum and retropleural space, which can be detected on CT scans. In previous reports, most hematomas due to parathyroid hemorrhage were located within the cervical region, and cases of spontaneous cervical-mediastinal and retropleural hematoma were extremely rare. Given the frequency of hyperparathyroidism in the general population and the few reported cases of hemorrhage complication, we conclude that extracapsular hemorrhage following a parathyroid adenoma is uncommon and may occur when the glands are enlarged secondary to adenomas.

Hematomas associated with parathyroid hemorrhage can require emergency surgery because of rapidly progressive airway and esophageal obstruction, hypercalcemia crisis, and acute blood loss. Conservative treatment with referral for elective parathyroid exploration is recommended for stable patients. In some cases, a delay of 1 month for the first surgical exploration is recommended [13-15]. Therefore, we performed surgery 1 month after presentation. Nevertheless, the surgical procedure was difficult because of tissue adherence and was further complicated by the hematoma. Considering the possibility of repeated hemorrhage, the patient should not be made to wait too long before surgical intervention [27]. The authors propose that surgical treatment should be delayed by 2 to 3 months after the cessation of bleeding. In case of an emergency, expedited surgical drainage can be performed using a minimally invasive approach. A well-ordered operation using a bilateral approach and intraoperative histologic examination can then be performed 3 months later [30].

The risk of parathyroid hemorrhage does not warrant surgery for asymptomatic hyperparathyroidism; however, during the long follow-up period, physicians should be aware of this serious and potentially fatal complication.

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